

allel in the respiratory sample, but in the literature it appears as the only microorganism isolated in culture that causes the disease.<sup>4</sup> *B. hinzii* also appears as a causal agent of bacteraemia in three cases described in the literature, with immunosuppression the common factor in two of them: one patient with myelodysplastic syndrome and another patient with HIV.<sup>4</sup> Similarly, *B. hinzii* also appeared as a causal agent of chronic cholangitis in a transplant patient on immunosuppressant treatment,<sup>5</sup> so it is considered that it may have a potentially pathogenic role in immunocompromised persons. There were no findings in our patient to suggest immunosuppression. As in some previous publications,<sup>2,3</sup> our patient had no known avian exposure, suggesting that the organism was obtained from another unidentified source. *B. hinzii* is usually resistant to, or has intermediate resistance to ampicillin, cefuroxime, ceftriaxone, cefotaxime, ciprofloxacin and tobramycin, and is sensitive to imipenem, meropenem, gentamicin, amikacin and trimetoprim–sulfametoxazol, which was similar to the antimicrobial sensitivity of our isolate, except as regards trimetoprim–sulfametoxazol. Molecular identification using MALDI-TOF-MS and 16S sequencing, as was performed here, provides the correct microbiological diagnosis.<sup>6</sup> The use of rapid techniques that increase the reliability and speed of identification of this microorganism could lead to clarifying its role as a coloniser and human pathogen.

## References

1. Cookson BT, Vandamme P, Carlson LC, Larson AM, Sheffield VL, Kersters K, et al. Bacteremia caused by a novel *Bordetella* species, *B. hinzii*. *J Clin Microbiol*. 1994;32:966–9.
2. Funke G, Hess T, von Gravenitz A, Vandamme P. Characteristics of *Bordetella hinzii* strains isolated from a cystic fibrosis patient over a 3-year period. *J Clin Microbiol*. 1996;34:966–9.
3. Gadea I, Cuenca-Estrella M, Benito N, Blanco A, Fernández-Guerrero ML, Valero-Guillén PL, et al. *Bordetella hinzii*, a “new” opportunistic pathogen to think about. *J Infect*. 2000;40:298–9.
4. Fry NK, Duncan J, Edwards MT, Tilley RE, Chitnavis D, Harman R, et al. A UK clinical isolate of *Bordetella hinzii* from a patient with myelodysplastic syndrome. *J Med Microbiol*. 2007;56:1700–3.
5. Arvand M, Feldhues R, Mieth M, Graus T, Bañadme P. Chronic cholangitis caused by *Bordetella hinzii* in a liver transplant recipient. *J Clin Microbiol*. 2004;42:2335–7.
6. Degand N, Carbonnelle E, Dauphin B, Beretti JL, le Bourgeois M, Sermet-Gaudelus I, et al. Matrix-assisted laser desorption ionization–time of flight mass spectrometry for identification of non-fermenting gram-negative bacilli isolated from cystic fibrosis patients. *J Clin Microbiol*. 2008;46:3361–7.

M. Pilar Palacián Ruiz,\* M. Alejandra Vasquez Martinez,  
Ana Isabel Lopez Calleja

Servicio de Microbiología, Hospital Miguel Servet, Zaragoza, Spain

\* Corresponding author.

E-mail address: [ppalacian@salud.aragon.es](mailto:ppalacian@salud.aragon.es) (M.P. Palacián Ruiz).

## Bilateral Pulmonary Sequestration in an Adult: Case Report and Review of the Literature\*

### Secuestro pulmonar bilateral en el adulto: aportación de un caso y revisión de la literatura

To the Editor,

We present the case of a 64-year-old man, ex-smoker, with clinical indicators of chronic bronchitis. He had been on home oxygen for more than 10 years and was on treatment for sleep apnoea-hypopnoea syndrome (SAHS) with nocturnal continuous positive airway pressure (CPAP). He was admitted in 1990 for pneumonia in the left lower lobe (LLL), secondary to symptoms of near-drowning in seawater. During the follow-up, triangular consolidation persisted in the LLL, but both computed tomography (CT) and fibrobronchoscopy failed to provide a diagnosis. CT-guided transthoracic needle aspiration was performed, with cytology negative for malignancy and a negative microbiological study. The patient was admitted on several occasions in recent years for exacerbation of COPD. A new chest CT scan was requested in 2004 due to consolidation in the LLL, and he was diagnosed with possible left intralobar pulmonary sequestration (PS). The patient refused any further studies. In 2009, he was admitted for pneumonia. The chest CT scan (Fig. 1) showed a 6–7-cm complex formation, in a medial location in the LLL, supplied by a large vessel originating in the inferior thoracic artery with drainage to pulmonary veins, corresponding to an intralobar PS with probable superinfection; in a symmetrical location, contralateral, there was another abnormal vessel, also originating in the aorta, corresponding to another intralobar PS in the right region. After improvement with treatment, he was discharged for follow-up at the clinic, but did not attend his check-ups. Most intralobar PS are unilateral, and bilateral

PS are very rare.<sup>1</sup> For Kohler, PS was first described by Rokitanski and Rektorzik in 1861, but it was not until 1946 when Pryce<sup>2</sup> made it known as a clinical entity. In 1972, Felson et al.<sup>3</sup> reported a case of bilateral PS confirmed by pathological examination. In 1977, Karp et al.<sup>1</sup> described another case in a 13-year-old girl who presented a mass in the LLL, discovered after a chest radiograph. CT scanning and pulmonary arteriography were carried out, showing a left intralobar PS, already suspected, and a right PS that was not previously observed. A few cases have been published since then, similar in that they concern paediatric or adolescent patients. In 2009, Yamamura et al.<sup>4</sup> explained the surgical procedure performed on a 44-year-old patient who presented bilateral intra- and extralobar PS. Wei and Li<sup>5</sup> analysed 2625 cases of PS in 2011, among which only 3 were bilateral and 2 of these intralobar. The case presented is doubly unique in that this is an older patient with bilateral intralobar PS. He presented recurrent pneumonia with a changing aspect in the LLL, in which diagnostic tests were inconclusive. The possi-



Fig. 1. CT angiography. The feeding arteries of both sequestrations, subsidiaries of the thoracic artery, can be seen.

\* Please cite this article as: Fontalba Navas M, et al. Secuestro pulmonar bilateral en el adulto: aportación de un caso y revisión de la literatura. *Arch Bronconeumol*. 2013;49:410–11.

bility of PS had already been described in the CT scan prior to his current admission. The existence of recurrent consolidations, especially if they occur in the LLL, requires us to consider the differential diagnosis of various conditions such as PS, long-standing pneumonia, chronic obstructive disease or lung tumours.<sup>6</sup> The diagnosis of PS has traditionally required pulmonary angiography to demonstrate abnormal vascularisation. However, new techniques such as next-generation CT angiography enable high resolution vascular reconstructions that could circumvent arteriography, as well as revealing congenital malformations, thereby avoiding more invasive techniques. With respect to treatment, we would propose acting on the left PS (as it is the symptomatic one) using surgery or VATS, the latter technique being less invasive.

## References

1. Karp W. Bilateral sequestration of the lung. *Am J Roentgenol.* 1977;128:513–5.
2. Pryce DM. Lower accessory pulmonary artery with intralobar sequestration of lung: report of seven cases. *J Pathol Bacteriol.* 1946;58:457–67.
3. Felson B. The many faces of pulmonary sequestration. *Semin Roentgenol.* 1972;7:3–16.
4. Yamamura Y, Hida Y, Kaga K, Kawada M, Niizeki H, Ichinokawa M, et al. Simultaneous resection of bilateral intralobar and extralobar pulmonary sequestration with video-assisted thoracoscopic surgery. *Ann Thorac Surg.* 2009;87:1939–41.
5. Wei Y, Li F. Pulmonary sequestration: a retrospective analysis of 2625 cases in China. *Eur J Cardiothorac Surg.* 2011;40:e39–42.
6. Palmowski M, Schreimer K, Hansmann J, Grenacher L. Bronchopulmonary sequestration: a differential diagnosis in young adults for recurrent pneumonia. *Lancet.* 2007;369:1318.

Montserrat Fontalba Navas,<sup>a,\*</sup> Justo Sánchez Gil,<sup>b</sup>  
José Calvo Bonachera<sup>c</sup>

<sup>a</sup> *Medicina de Familia, Complejo Hospitalario Torrecárdenas, Almería, Spain*

<sup>b</sup> *Unidad de Gestión Clínica de Medicina Interna, Hospital Universitario Reina Sofía, Córdoba, Spain*

<sup>c</sup> *Unidad de Gestión Clínica de Neumología, Complejo Hospitalario Torrecárdenas, Almería, Spain*

\* Corresponding author.

E-mail address: [mnfont@hotmail.com](mailto:mnfont@hotmail.com) (M. Fontalba Navas).

## Obliterating Bronchiolitis in a Patient Treated With D-Penicillamine<sup>☆</sup>

### *Bronquiolitis obliterante en paciente tratado con D-penicilamina*

To the Editor,

Various side effects during long-term penicillamine treatment have been described, especially dermatological reactions, such as exfoliative dermatitis, rash and alopecia.<sup>1</sup> Other adverse events, such as vasculitis and thyroiditis, have also been reported. Respiratory effects have been documented, albeit less frequently, including cases of asthma, pulmonary fibrosis, interstitial pneumonitis and bronchiolitis obliterans.

We present the case of a male patient, 47 years of age, never-smoker, with a clinical history of hypertension on medical treatment, dyslipidemia on treatment with statins, Graves-Basedow disease treated 8 years ago with radioiodine (currently receiving replacement therapy) and Wilson's disease diagnosed 20 years ago, initially treated with zinc and for the past 2 years with penicillamine. He had a work history as a salesman, with no contact with toxic substances or poultry. The patient consulted due to dyspnea on exertion. An initial lung function study was performed which showed severe airflow limitation, so treatment with inhaled corticosteroids was initiated. A computed tomography (CT) scan of the chest was requested, along with complete lung function testing, revealing severe irreversible obstructive changes with air trapping and reduced carbon monoxide diffusing capacity (DLCO). Discrete bronchiectasis and diffuse bronchiolectasis were observed on the CT scan (Fig. 1). A sweat test to rule out cystic fibrosis was negative and an immunoglobulin study was normal. During the follow-up visit, respiratory failure was observed, so the decision was taken to admit the patient for further tests. On physical examination, oxygen saturation (breathing room air) was remarkable due to pulse oximetry of 88% without signs of labored breathing and normal pulmonary auscultation. The rest of the physical

examination was normal. General clinical laboratory tests were unchanged and the chest CT was the same as before. In view of suspected drug-induced bronchiolitis, a biopsy was obtained from the lingula by anterior thoracotomy. After an uneventful post-operative period, the patient was discharged with domiciliary oxygen therapy. The final diagnosis from the pathology study was follicular bronchiolitis associated with constrictive bronchiolitis obliterans.

The decision was taken to discontinue penicillamine treatment and start treatment with bronchodilators and corticosteroids (methylprednisolone 40 mg/day), and the patient was referred to a reference center for evaluation for lung transplantation. In May 2012, a double-lung transplant was successfully performed.

Bronchiolitis obliterans is a non-specific disorder of the small airways (<2 mm in diameter). Its clinical presentation is very non-specific (cough and progressive dyspnea). Physical examination is unremarkable and signs of hyperinflation, prolonged expiration and non-specific breathing noises such as rhonchi, crackles or wheezing may be observed. A plain chest X-ray may be normal or show signs of air trapping. Additional information can be obtained from a high resolution CT scan during inspiration and expiration. Distinctive radiological signs in the expiration slices have been described on the CT scan which could guide diagnosis, whether direct (bronchiolar wall thickening with the typical "tree-in-bud" pattern, bronchiolectasis and centrilobar nodules) or indirect (subsegmental atelectasis and signs of air trapping). This syndrome can be classified by etiology or pathology.<sup>1,2</sup> From an etiological point of view, idiopathic cases must be distinguished from those caused by secondary factors. These latter cases are usually acute, the causative agent (primarily drugs or toxic substances) is known, and there is a good response to bronchodilator treatment. The following histopathological classifications have been described: constrictive, proliferative and follicular bronchiolitis, central interstitial fibrosis and diffuse panbronchiolitis. Bronchiolitis can be diagnosed from the clinical signs and symptoms and appropriate complementary tests, but the definitive diagnosis is obtained from lung biopsy. Treatment is based on corticosteroid therapy, bronchodilators, macrolide antibiotic treatment and in the case of rapid progression, lung transplant.

Our patient presented a case of constrictive bronchiolitis obliterans which has an uncommon histological pattern, characterized

<sup>☆</sup> Please cite this article as: Bruguera-Àvila N, et al. Bronquiolitis obliterante en paciente tratado con D-penicilamina. *Arch Bronconeumol.* 2013;49:411–12.